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[CASE REPORT]

Electroconvulsive Therapy for Long-term Mutism in a Case of Noncatatonic Paranoid Schizophrenia

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ABSTRACT

We report a rare case of paranoid schizophrenia presenting with continuous mutism for about three years. This 26-year-old woman with multiple Schneiderian first-rank symptoms ['Schneiderian' refers to those symptoms established by the German psychiatrist Kurt Schneider for the diagnosis of schizophrenia] did not have any catatonic features, and she would fluently communicate by gesturing or writing. Since there was serious impairment in biological functions not readily correctable by antipsychotics, she was started on electroconvulsive therapy. She responded well to 14 sessions of electroconvulsive therapy along with oral haloperidol. We also discuss the cultural implications of prolonged mutism. To the best of our knowledge, this is the first case of mutism in noncatatonic paranoid schizophrenia that responded well to electroconvulsive therapy described in the literature.

INTRODUCTION

Mutism has been defined as functional inhibition of speech and vocalization. Rarely an isolated disability, in psychiatric literature it is accompanied by disorders in behavior, motor symptoms, and disturbance of affect and thought. The most common motor and behavioral dysfunction to be associated with mutism is catatonia.[1] Several psychiatric disorders, including schizophrenia, mania, depression, and dissociative disorder, are known to cause mutism. Important organic causes of mutism are dementia, neurodegenerative and demyelinating disorders, head injury, posterior fossa surgery, encephalitis, frontal lobe lesions, postictal phase of epilepsy, laryngeal tumors, and endocrine disorders (e.g., hyperparathyroidism, myxoedema, diabetic ketoacidosis Addison's disease).[2] Medications capable of inducing mutism include immunosuppressants such as cyclosporine,[3] maprotiline hydrochloride, and aspirin intoxication. Among substance-related conditions, phencyclidine and alcohol-induced psychosis can cause mutism. In childhood, important causes of muteness are deafness, developmental failures, autism, elective mutism, and conversion disorder.

In association with schizophrenia, mutism is rare in developed countries and mostly associated with catatonia.

In patients with noncatatonic schizophrenia, only a few cases were found in the literature. Though the time length to qualify for prolonged mutism has not been formally defined, mutism extending up to several years has been found among inhabitants of the Micronesian island of Kosrae, where one patient was mute for 20 years and another for three years.[4] Apart from these cases, there are two reports of prolonged mutism in noncatatonic schizophrenia in the adult population.[2,5] Another case of childhood onset noncatatonic schizophrenia of duration 17 years, resistant to treatment, has been described by Altshuler et al.[6] In their paper, Altshuler et al. do not mention duration or treatment of mutism.[2] Other cases have been managed with antipsychotics. We describe a case of paranoid schizophrenia without any catatonic signs and symptoms presenting with long-term mutism that was successfully treated with electroconvulsive therapy (ECT). To the best of our knowledge, this is the first such case described in the literature.

CASE REPORT

Ms. B, a 26-year-old woman, presented to our clinic with insidious onset psychiatric illness of about five years duration. She was educated until intermediate level and belonged to a low socioeconomic status Hindu joint family of urban background. She had no past or family history of psychiatric illness, and she had good premorbid functioning. Her symptoms started with academic decline, suspiciousness, and referential ideation progressing to gradual social withdrawal, predominant irritable mood, and delusion of persecution by her college authorities and employer. A year after these initial symptoms appeared, she began to exhibit odd behavior such as sewing clothes in strange patterns, offering worship by performing strange rituals, and riding a tireless bicycle at midnight for no apparent reason. She had disturbing auditory hallucinations that were commanding type and voices commenting on her actions. Auditory hallucinations were followed by visual hallucinations in which she would see a man and woman entering inside her body and controlling movements against her will. She would argue with her family members that in order to accommodate them, her body had grown physically. She believed this man and woman were preparing photographs and film rolls inside her body, and to expel them she would slap herself repeatedly. She would often stay awake at night fearing that they would perform some foul play with her body. However, it could not be ascertained retrospectively whether she would simultaneously have any bodily sensations. During this period she would often undertake 'maun-vrat,' a time-limited voluntary mutism that is culturally and religiously sanctioned in India for spiritual growth, for few days at a stretch, communicating through gestures and writings only. Initially, she would claim to do so for the well-being of herself and her family.

During the third year of her illness, she developed persecutory delusions against family members—that they were conspiring to take her property. She alleged they poisoned her, causing her to lose her voice. Hence, for the last three years, she completely stopped speaking and would communicate everything by gesturing or by writing.

Thereafter, she deteriorated in that she was irritable, had impaired self-care, slept very little, and ate minimal amounts of food because of fear of harm by family members. About six months before admission she lodged a complaint with the police and filed a court-case against them. The legal services asked her to get a psychiatric assessment performed, which led her to approach our psychiatric outpatient services where she was promptly admitted. There is no history of jaundice, organicity, substance use, or sexual exposure.

After admission, her initial general physical examination and investigations were within normal limits. Most of her higher mental functions were intact (Mini-Mental State Examination [MMSE]=28) but she did have impaired judgment, abstraction, and absent insight. In the absence of prominent mood symptoms, organicity, and substance use, and based upon several Schneiderian first-rank symptoms ('Schneiderian' refers to those symptoms established by the German psychiatrist Kurt Schneider for the diagnosis of schizophrenia), her bizarre delusions, and her lack of prominent disorganization, catatonic, and negative symptoms, she was diagnosed as paranoid schizophrenia (F20.0, as per *International Classification of Diseases, 10th Revision*). On admission, her Positive and Negative Syndrome Scale (PANSS) score was 94 (positive=34, negative=30, general psychopathology=30). She was started on olanzapine tablets, which were increased to 20mg over the next two weeks, but there was no response. She was not willing to accept any other medication or solid food, so it was decided to apply electroconvulsive therapy (ECT) after a valid informed consent from her legal guardian. At our center, modified bilateral ECT is administered using an indigenously manufactured brief-pulse, constant-energy machine (Medicaid Systems, Chandigarh, India) three times weekly, and during the procedure, the duration of current is varied while keeping the frequency at 70Hz and the pulse width constant at one millisecond. Atropine is used as a premedication, thiopental sodium for induction, succinylcholine is used for muscle relaxation, and EEG monitoring is done. Our patient was given 14 ECT treatments (Table 1), all of which produced generalized seizures for adequate duration, and she recovered each time uneventfully. She started improving after three ECT treatments in terms of biological functions, uttering proper audible words after nine ECT treatments, and speaking as usual after 12 ECT treatments. She was also started on haloperidol tablets, titrating to 30mg after discontinuing the olanzapine. At the time of discharge, the PANSS score improved to 44 (positive symptoms=14, negative=12, general psychopathology=18), as well as her

TABLE 1. Serial electroconvulsive therapy (ECT) and clinical parameters

ECT TREATMENT NO.	WEEKLY PANSS SCORE	TRAJECTORY OF IMPROVEMENT
1	94	Baseline score
4	82	Biological functions improved
7	65	Persecution improved
9	60	Speaks after three years
12	44	Speaks as usual self
14	44	Discharge

body mass index (BMI) (previously 15.44kg/m^2 and now 20.76kg/m^2) and Global Assessment of Functioning (GAF) score (from 25 to 55). She did, however, only have partial insight during discharge, and long-term follow-up along with proper psychoeducation of both patient and caregiver was planned.

DISCUSSION

Mutism should be carefully evaluated because of its varied causes. Clues to neurological causes include irregular respiration, abnormal pupil responses, roving eye movements, facial weakness, and exaggerated jaw jerk. On the contrary, patients with primary psychiatric disorders may be induced to whisper or communicate in writing, although the latter may also occur with infarction, leading to pure word-dumbness (an apraxia restricted to the movements required for speech). Our patient did not have any focal neurological deficit and her associated paranoia, first rank symptoms, and bizarre delusions unequivocally established paranoid schizophrenia as diagnosis. We ruled out catatonia by applying the Bush Francis Catatonia Screening Instrument (BFCSI), according to which catatonia should have at least another sign/symptom unlike our case.[7] Neither could the mutism be explained by negative symptoms on account of her elaborate and spontaneous self-expression by writing and absence of other severe negative symptoms.

From the management perspective, since our patient had not accepted any solid food for 3 to 4 weeks before admission and the very nature of her psychopathology, due to which she was reluctant to take medications, we were impelled to make an early decision in the favor of ECT treatment, as we believed in this case a quick and effective intervention was imperative. ECT is a very safe procedure with no recorded history of related mortality or severe adverse effect in our center. In fact, this patient had no adverse effects or cognitive decline (as recorded by MMSE) after 14 consecutive ECT treatments.

'Maun-vrat' is a culturally and religiously sanctioned practice of mutism in India. It is used as a coping response to social stressors, very much like with the Kosraeans. However, in our case, the practice, though ratified by the family initially, was later disapproved because the religious practice is time-bound and goal-directed.

Prolonged mutism in schizophrenia is currently rare in Western literature and is mostly associated with catatonia. But it was quite common in the pre-neuroleptic era and was the primary clinical correlate of the length of untreated illness.[8] The fact remains that in the last decade both the reported cases were from India and had drug-resistance.[5,6] This is quite significant in view of the above socio-cultural and religious practices. At least in Kosraeans, those with noncatatonic schizophrenia commonly present with mutism in the early course of illness as a culturally mediated expression of psychopathology, which typically responds well to antipsychotics.[4] It is not clear whether the reverse is true for Indian culture (i.e., long-term mutism in the late part of illness indicating difficult course). Alternatively, it may simply be the result of scarce mental health resources, which increase the duration of untreated psychosis as in the pre-neuroleptic era in western countries. These unanswered questions can only be addressed when we have large-scale studies testing specifically these hypotheses given the rarity of this condition in any population.

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